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the description of two cases,
Mother and Daughter, by J. W.
Ballantyne, M. D., and George
Elder, M. B., C. M., Edinburgh.



PEDIATRICS



Original Articles.

TYLOSIS PALMAE ET PLANTAE: WITH THE DESCRIPTION OF TWO CASES, MOTHER AND DAUGHTER.

By J. W. BALLANTYNE, M.D., and GEORGE ELDER, M.B., C.M.

Edinburgh.

AMONG the various forms of epidermal disorder which have hyperkeratosis as their evident feature *tylosis palmarum et plantarum* is specially worthy of note. This disease may be defined as a hypertrophy of the horny layer of the epidermis affecting only the palms of the hands and the soles of the feet and leading to the development of a hard plate in those regions.* The case about to be described, which was carefully examined by both of us, occurred in Dr. Elder's practice; the history of the patient and her family will now be given and thereafter the cutaneous affection itself will be more fully considered.

DESCRIPTION OF A CASE OF TYLOSIS PALMAE ET PLANTAE.

The patient, E. C., a girl eight years of age, shows a distinct callosity of the palms of the hands and of the soles of the feet. The condition is most noticeable in the palms, to which it gives a peculiar yellow color, and a hardness and roughness readily noticed on shaking hands. The thickening resembles that present on the hands of a workman who uses tools frequently, and is all the more prominent from its absence at the flexures. It is greatest on the hypothenar eminences, but it is also present on the thenar eminences and along the palmar aspect of each finger, as well as over the proximal part of the palm. Indeed no part of the palm is quite free from it, save at the lines of flexure. It is strictly limited to the palm and does not reach the dorsum of any of the fingers, and above it is sharply defined by the line of flexure of the wrist, beyond which there is absolutely no trace of it. The distribution of the thickening is shown by the accompanying figure, in which the thickening is represented by shading—the intensity of the shading corresponding to the extent of the thickening.

* Crocker (H. R.) *Diseases of the Skin*, 2nd Edit., p. 363, 1893.



The feet are also affected in a similar manner, although here the thickening is not quite so noticeable. It is most marked along the lines of pressure of the sole, and as in the hands it is strictly limited to the palms, so here it does not extend beyond the soles.

The condition was first noticed by the mother when the girl began to walk about freely, and it has remained present ever since, even when there is practically no friction on the hands. It is, how-



ever, aggravated by friction, and the right hand, which is more used than the left, shows a greater degree of thickening. Her mother states that the hands and feet "peel" in pretty large scales at times, usually every spring and autumn; but even when this occurs the skin beneath is still distinctly affected. A few weeks ago, after

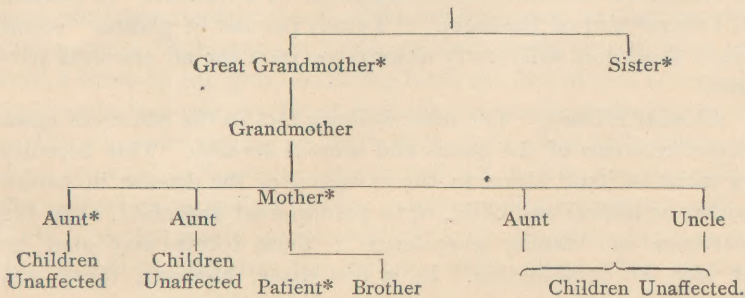
scarlet fever, there was skinning of the palms and soles, the scales shed being very large and thick. Before the skinning commenced the palms were much thicker and yellower than usual.

When the patient went first to school she developed a large number of warts on the fingers, said to have been acquired from the girl who sat next to her in class ; but at present scarcely any of these growths remain. No other peculiarity of the skin can be observed. The teeth, hair, and nails seemed quite normal.

The point of special interest, however, in the case is the hereditary history. Her mother has a similar condition of the palms and soles, noticed soon after birth, and all her life her rough hands have been a source of some annoyance to her. She has also constantly to use a file to keep in check the growth of the skin at the roots of the nails. The patient's father's hands are quite free, as are also those of an only brother, who is considerably over one year in age. No other members of the family could be examined ; but it was elicited that none of the father's family had ever been affected, whilst the following interesting history was got from the mother, a particularly reliable and intelligent lady.

She, the girl's mother, is French, and, as already mentioned, has "hard hands" like those of the patient. Of her sisters, three in number, one has a similar condition ; the other two have not, and neither has her brother. The maternal grandmother of the patient had nothing of the kind ; but the great grandmother (maternal), as well as her sister, had a similar condition. No further particulars could be elicited save the remark that there was a tradition that it had been in the family for generations, but that it affected only female members and only some of them ; that it was only transmitted through the female members, and that it usually skipped a generation. In the present instance, however, this last peculiarity is absent, for both mother and daughter are affected.

For the sake of clearness this history may be represented as a



genealogical tree, in so far as it is known to the patient's mother. In this the persons affected are indicated by a *.

For years the patient's mother has been using glycerine and pumice stone for the thickening; but although the condition was thus made less distinct it was by no means removed. A few weeks ago a solution of salicylic acid in ether (10 grs. to the oz.) was ordered to be applied, and the state of the hands has been already much improved; but, of course, it would be rash to expect a complete cure.

TYLOSIS PALMAE ET PLANTAE.

Historical Notes. According to Unna (7),* who described the disease with some fullness in 1883, Ernst (1) and G. Simon (*Die Hautkrankheiten*, p. 48, 1851) were the first to record cases. In 1851, also, Pickells (2) noted what he regarded as a congenital ("connate") instance of it in a boy of 13 or 14 years of age, and in 1879 Bulkley (3) saw a woman of 25 in whom it had been present "since infancy." Thost's inaugural dissertation (4) contained a most interesting account of a family in which this disease was hereditary. Other cases were those noted by Bocgehold (5) and Startin (6). Since Unna's classical paper appeared in 1883, G. H. Fox (8), Biart (9), Date (10), Jonathan Hutchinson (11), Crocker (12), H. G. Brooke (13), Sherwell (17), Bassaget (18), Dupré and Mosny (19), Hallopeau (20), and Bergh (21) have all written on the subject and have added to the list of known cases.

Synonyms. As with so many dermatological states so with tylosis palmarum et plantarum a number of synonymous terms are in use. It is the "ichthyosis palmaris et plantaris" of Thost and others, the "tylosis palmarum manus plana" of Hebra, the "keratoma plantare et palmare hereditarium" of P. G. Unna, the "inherited keratosis of the palms and soles" of Hutchinson and G. H. Fox, the "hyperkératose palmaire et plantaire" of Hallopeau, and the "kératodermie symétrique des extrémités congénitale et héréditaire" of Besnier and Doyon* and of Bassaget. "Tylosis palmarum et plantarum" seems to us a short and sufficiently descriptive term for all practical purposes.

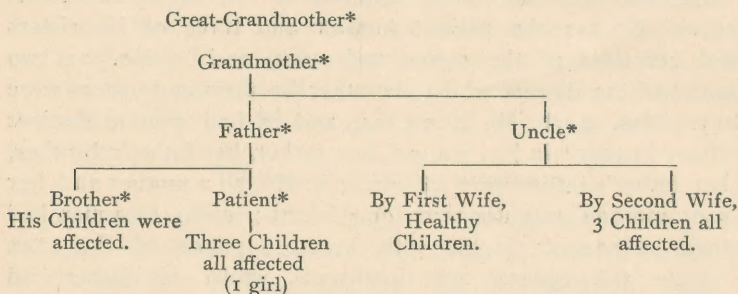
Clinical History. The most striking fact in the history of cases of hyperkeratosis of the palms and soles is *heredity*. This heredity may manifest itself either in the existence of the disease in parent (mother or father) and child, or in grandparent and child, or in the occurrence of "family prevalence"; these forms may also be met with in combination. In a few cases, however, there was

*The figures within brackets refer to the bibliography at the end of the article.

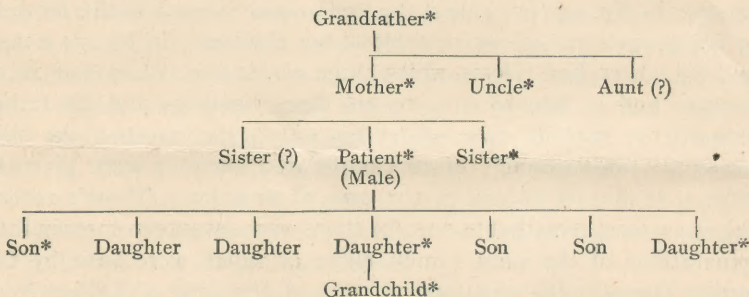
either no hereditary history or no reference to it, viz. in Nos. 5, 6, 17, 19 and 20. The least marked form of heredity—family prevalence—was present in Nos. 2, 11 and 21; in No. 2 the boy described and the other children of the family were affected, in No. 11 the patient himself and three of his sisters showed keratosis of the palms and soles, and in No. 21 two brothers had the disease whilst six other brothers and sisters were healthy. Nos. 3, 7b, 8b, 9, 13, 14, and 18 had a more distinct hereditary history; in No. 3 a girl, her father, her father's brother, and her father's father were affected; in No. 7b a mother and her two sons and one daughter were the subjects; in No. 8b a man had keratosis palmarum et plantarum with keratosis pilaris of the arms and thighs and general mild ichthyosis, whilst his father had the palmar and plantar lesions; in No. 9 a man, his brothers and sisters, his father and his father's brother were all sufferers; in No. 13 (two cases) the lesion was present in the mother, in her parents and in one or more of her children; in No. 14 a man and his father had tylosis, whilst three of his five sisters had xeroderma; and in No. 18 a man, his three brothers and his father showed the morbid state, whilst his only sister escaped the skin lesion, but died insane. More complicated histories were given in Nos. 4, 7a, 8a, 10, 12 and in our own observation. Thost's record (4) was a most remarkable one, for there were seventeen cases in four generations of the same family, eight of which were seen by the author himself: the great grandfather of the little girl (Lina Neubrandt) had keratosis palmarum et plantarum; he had seven children, six males and one female, of whom five (all males) inherited the disease; of the five affected males, one died young, one was lost sight of, two had families in which there was at least one case of the malady, and the fifth (Heinrich Heilmann) was twice married and had a large family, consisting of eight children, five males and three females, of whom six (four males and two females) were similarly affected; finally Heinrich Heilmann's eldest daughter (Margarethe Neubrandt) had four children, three girls and one boy, and of these all the girls, including Lina, the first of this remarkable family who was seen by Thost, had the cutaneous anomaly. In one of Unna's observations (7a) there were eight cases (five males and three females) in three generations and the malady was transmitted through males as well as females; and one of the patients seen by G. H. Fox (8a) was a member of a family of eight, every second one of whom inherited the disease, while his mother, mother's

* Besnier (E.) and Doyon (A.) *Maladies de la peau* (Kaposi), Vol. II., p. 41. 1891.

father, an uncle, and two nephews were similarly affected. The genealogies of Nos. 10 and 12 may be given in tabular form. That of Horton Date's patient is as follows :



The family tree of Crocker's patient (12) was very interesting.



In this case, which closes this remarkable demonstration of heredity, nine individuals in five generations were certainly affected with the disease, four were not so affected, and with regard to two there was some doubt. From what has been said it will have been learned that when the heredity was direct the transmission was sometimes through the males only (Nos. 3, 8b, 9, 14 and 18), less often through the females only (Nos. 7b, 13 and 22), and sometimes through the males of one generation and the females of another (Nos. 4, 7a, 8a, 10 and 12). In our case (22) the females only were affected, and skipping a generation had occurred at any rate once.

There is no limitation in the matter of *sex*, for an analysis of the family histories shows that out of ninety-three cases in which the sex was stated fifty-seven (61 per cent) were males and thirty-six (nearly 39 per cent) were females. There is thus a distinct preference for the male sex. Possibly the fact that men are more

actively engaged in manual labor than women may serve to account for the above circumstance; their environment is more favorable to the development of the latent and hereditary tendency.

With regard to all the cases which have been referred to in this paper there is good reason for supposing that they were congenital.

We do not mean to say that in all of them was the anomalous condition of the epidermis of the palms and soles present at birth, although in some instances this was affirmed, but on hearsay evidence. In all, however, it would seem that the predisposition to the disease was truly congenital, although pressure and friction might be necessary to develop the characteristic lesion. In all Thost's cases (4) it was said that the disease was observed in the first week of life as a slight roughness of the epidermis of the palms and soles, which rapidly increased in thickness. With regard to the second family seen by Unna (7b), the interesting remark is made that immediately after birth both palms and soles were observed to be surrounded by a bluish border, on which, after the seventh month horny growths began to appear, and from which these growths soon spread to the rest of the palmar and plantar surfaces. In other cases the lesion was first noticed at three months, or when the child began to creep, or when it began to walk (e.g. in No. 22); and in one instance (No. 20) it would seem that it became evident only at the age of 20 years, nevertheless Hallopeau, the reporter, regards it in this instance also as of embryonic origin. It may be concluded that if the skin of the palms and soles were carefully examined at birth some slight morbid change would be discovered in some cases at least.

Symptomatology.—In all the recorded cases both the palms of the hands and the soles of the feet showed thickening of the epidermis, and it is from this typical distribution of the lesion that the disease has got its name. In Nos. 18, 19 and 21 the hyperkeratosis was not limited absolutely to the palmar and plantar surfaces, but affected to some extent the dorsal surface of the digits also, in Nos. 2 and 5 a similar condition was found on the knees; and in Nos. 11 and 14 there was general xeroderma. Nos. 8b and 19 were interesting: in the former there was keratosis pilaris of the arms and thighs, general ichthyosis of a mild type, and tylosis palmarum et plantarum; whilst in the latter the palmar and plantar lesion was accompanied by a similar epidermic thickening over the patella and olecranon and on the lips and mucous membrane of the palate, as well as by mild ichthyosis of the extensor surfaces.

With regard to the characters of the lesion itself, the descrip-

tion which we have given in connection with the girl, E. C., may be taken as typical of the minor degree of the malady. The plantar and palmar surfaces of the feet and hands are covered by a thick yellow or yellowish-brown cuticle like a layer of wax. This gives to the palms and soles a peculiar hard and horny feeling. Sometimes the cutaneous thickening is greater over the thenar eminence, sometimes over the hypothenar, but as a general rule it is most marked along the lines where there has been most pressure. In more marked cases the thickened epidermis is split up by grooves or fissures into a large number of irregular areas or islands and the whole forms a mosaic. The grooves vary in depth and in width and have no connection with the natural lines of flexure of the palms and soles, in some instances, indeed, crossing them at right angles. In parts subjected to much pressure the fissures may disappear altogether. Every now and again desquamation of the thickened cuticle takes place, sometimes, as in our case, in spring and autumn, in other instances in the autumn only, as in No. 12.

The lesion is usually painless. If the hypertrophied epidermis be removed, a red, bleeding surface with enlarged papillæ is exposed. There is often some difficulty in completely flexing the joints of the palms and soles. The affected area is not infrequently surrounded by a narrow erythematous border or zone, which passes insensibly into the surrounding healthy skin. The condition becomes more marked as age advances up to a certain limit; it is thereafter practically permanent, or is kept in check by the patient use of pumice-stone, etc.

In certain of the recorded cases itching was a prominent symptom. Tactile sensibility is present, but in a blunted state; and sensibility to heat and cold, and also to pain, is usually much diminished. In most cases there is a dryness of the affected skin (xeroderma); but in others (Nos. 7 and 18) there was hypersecretion. In No. 7 also the sensibility to touch was unimpaired, and there was osmidrosis as well as hyperidrosis. The nails have been found in a ragged state and incurved on themselves; in No. 20 there was a certain degree of sub-ungual parakeratosis.

The condition of the hair and teeth is not often reported; but in No. 18 both these structures were normal. The general health is usually quite good, although in one or two cases (Nos. 18 and 25) coldness of the extremities and "dead fingers" were complained of, and in No. 3 there was rheumatism. Sometimes a neurotic family history has been recorded, but the patient is generally free from nervous symptoms and visceral lesions.

Pathology.—The horny layer on the palms and soles had a thickness, varying in most cases, from one-eighth to one-sixteenth of an inch; but over the ball of the thumb or great toe it might reach half an inch. The other naked-eye characters of the lesion have been referred to under the symptomatology; but it may be noted that Hallopeau was of opinion that in the case observed by him (No. 20) the epidermic thickening was especially marked round the dilated sudoriparous gland orifices. Thost (4) has described the microscopic appearances of the diseased integument as follows: The papillae were increased in length five-fold, their breadth was somewhat less than normal, and their number was not greater than usual. The prickle-cells were not enlarged or otherwise altered; but their number was greatly increased, and on this account the rete malpighii was much broader. The stratum granulosum was normal. The horny layer was much thicker than in the healthy condition, and the increase was chiefly in its middle layer. The cutis, as a whole, was thicker than usual, and the adipose tissue more marked. The blood-vessels appeared to be enlarged. A general hypertrophy of all the layers of the skin was noted by E. Dupré and Mosny (19).

Pathogenesis.—The names which have been given to this disease by various writers indicate that all are not agreed as to the nature of the morbid process. Unna (7) places it in the group of the acanthoses along with condyloma acuminatum, verruca, fibroacanthoma cornificans, tyloma, and clavus, looking upon it, indeed, as a variety of cornu cutaneum; he gives ten reasons why it should be regarded as more nearly allied to horns than to ichthyosis. Bronson* adopts Unna's view of the matter. Others have regarded it as a localized form of ichthyosis, and that it is closely related to the condition known as fetal ichthyosis is beyond doubt. Whether, however, it is a pure hyperkeratosis or is both a hyperkeratosis and a hyperacanthosis may in the meantime be left unsettled, for even as regards foetal ichthyosis this question has not been decided. Hallopeau (20), it may be remarked, regards it as a form of naevus and as of embryonic origin; but *naevus* can scarcely be looked upon as a precise pathological term. Dupré and Mosny (19) call it a congenital lesion of the skin of an ichthyotic kind, in certain points resembling congenital verrucose naevi. Whatever name be given to the lesion it is essentially a hyperplasia of the epidermis on certain clearly defined areas of the surface of the body.

With regard to its mode of origin also opinions differ. That it is congenitally pre-disposed to seems, however, to be quite clear; it

* Bronson, E. B., Journ. of Cutan. and Ven. Dis., ii, p. 206, 1884.

also appears to be certain that this pre-disposition is rarely strong enough to bring about the full evolution of the disease, and that an exciting factor, in the form of intermittent pressure or some other irritant is necessary to develop the characteristic lesion. This view is supported by the markedly hereditary character of the malady, to which reference has already been made. Hutchinson (16) says with regard to one of his cases: "No doubt the child was born with a peculiarity in the structure of the skin which rendered it less capable than usual of resisting the effects of mechanical irritation; thus when the child began to walk and to handle toys the skin revealed its defective organization by passing into a condition of chronic inflammation. In this respect we have a parallel with certain other well-known types of disease, such as retinitis pigmentosa, Kaposi's disease, and many others in which nothing is to be observed at the time of birth, but in which exposure to light, sun, etc., at once brings on a peculiar form of degeneration of tissue." What this congenital peculiarity in the structure of the skin is cannot be certainly determined; but that it is in some way connected with an unusual state of the epitrichium is very probable. Ohmann-Dumesnil* suggests that the permanent adherence of this embryonic layer is the cause of the various thickenings, general or localized, to which the name of ichthyosis has been given. This may be so; but it is well to remember that the normal functions of the epitrichium are not yet known, and that speculations as to its pathology are, therefore, premature. Still the suggestion is a valuable one, and it may be that in the epitrichium and its morbid states may be found the causes of many, if not of all, congenital epidermic anomalies.

Brooke (13) is of opinion that the symmetry of the lesions is not sufficient ground for supposing a primary central causation; some cases are to all appearances primarily central tropho-neuroses, but in the majority the fact that it spreads from one limb to another or from hands to feet, or *vice versa*, supports the theory of a reflex origin. Hallopeau's observation (20) that the morbid process begins round the dilated orifices of the sweat glands is of interest; this peculiarity is shared by other skin diseases, e. g., pityriasis rubra pilaris, seborrhoeic eczemas, lichen planus, etc.

We have thus far been speaking only of cases in which tylosis palmarum et plantarum was congenital in origin in at any rate the wide sense of the word; but an apparently identical lesion may be produced in adult life as a consequence of the administration of arsenic. Recent instances of this condition have been reported by A.

*Dumesnil (A. H. Ohmann). "The human epitrichium." *Teratologia*, ii, p. 155, 1895.

Mathieu* and A. Giletti** (although Giletti doubts the arsenical origin of his case). Even in these cases, however, a congenital predisposition may have been present, and the arsenic may have acted as the exciting and not as the sole cause.

A somewhat similar malady has been described by Max Neuburger (*Monatshefte f. prakt. Dermat.*, xiii, p. 1, 1891) under the name of akrokeratoma hereditarium, but in it the extensor surfaces and especially the backs of the hands and feet were the parts affected, and the lesion, although resembling in its nature that in tylosis, was not sharply limited in distribution.

Treatment.—The usual treatment adopted in cases of tylosis palmarum et plantarum is the purely palliative one of applying glycerine to soften and using pumice-stone to remove the hardened and thickened epidermis. "In congenital cases," says Crocker, "a cure can, *a priori*, scarcely be expected," yet in our case distinct improvement has already followed the painting of the affected parts with a solution of salicylic acid in ether, and Unna (7) cured five members of one family with a similar but stronger solution (10 per cent.) Ichthyol, also, has been recommended; Bulkley (3) employed diachylon ointment; and Hutchinson (14) advised arsenic, tar ointments, and the use of cork soles. We are not aware whether thyroid feeding has yet been tried in such cases.

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22. Our own observation, as related above.

NOTE.—To Ernst's paper we have been unable to refer in the original ; all the other references have been verified.

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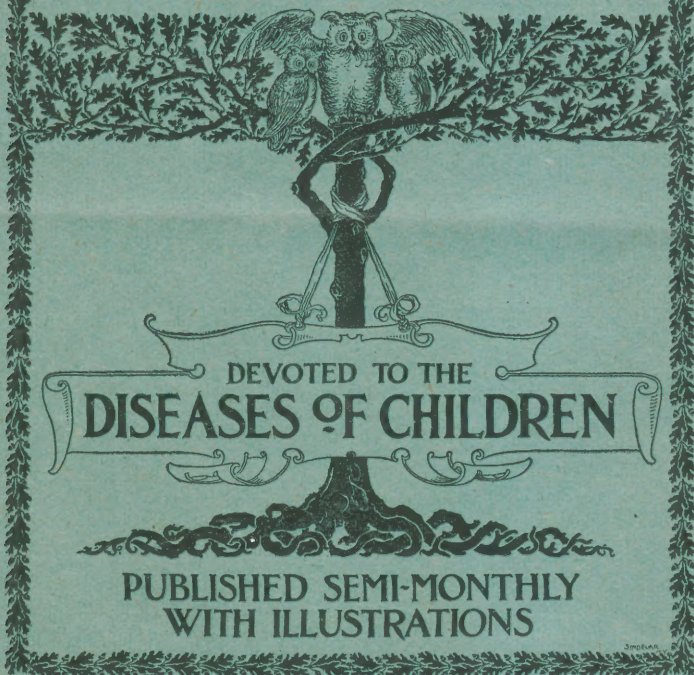
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